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Congenital Pulmonary Malformations Diagnosed Over a Period of 10 Years☆

Malformaciones congénitas pulmonares diagnosticadas en un periodo de 10 años

To the Editor,

Congenital lung malformations (CLM) are a group of entities caused by alterations in the embryogenesis of the lung and airways. Both the site within the tracheobronchial tree and the gestational age at the time of the embryological insult correlate with lesion type and histopathology.[1]

Clinical, radiological and histopathological criteria have been established for the classification of most CLM. Diseases currently considered CLM are congenital pulmonary airway malformations (CPAM), pulmonary sequestration, bronchogenic cyst, congenital lobar emphysema, and bronchial atresia. Stocker[2] classifies CPAM, formerly known as congenital cystic adenomatoid malformation, into 5 subtypes based on the number and size of the cysts and their anatomical origin: Type 0: lung consisting of small cysts (0.5 cm), incompatible with life; Type 1: single or multiple cysts (2–10 cm), arising from the bronchi or bronchioles; Type 2: multiple cysts (0.5–2 cm) in the bronchioles; Type 3: solid lesion with cysts (<0.5 cm) in the bronchioles and alveolar tract; and Type 4: cysts up to 7 cm, originating in the distal acinar region.

Pulmonary sequestration refers to a non-functioning sector of the lung that is irrigated by the systemic circulation, and is classified as intralobar or extralobar, depending on whether it is contained within the visceral pleura of lung or it has its own pleural lining, respectively.[1]

CLMs are rare, accounting for between 7.5% and 18.7% of all organ malformation, and their clinical presentation and severity vary widely, especially in terms of the degree of pulmonary involvement. Manifestations can occur at any age, but typically develop in infancy and childhood. The risk of recurrent respiratory infections or malignant transformation has been described in the course of some CLM, particularly CPAM.[3,4]

CLM may be diagnosed at birth due to the onset of clinical symptoms, incidentally during radiological studies, with or without symptoms, or in ultrasonography tests performed in the prenatal period.[1,4,5]

The management of these lesions depends on the type of malformation and the development of symptoms, so management must be individualized for each case and type of malformation; most authors recommend resection of the lesion, but there is currently no consensus on the surgical approach, especially in asymptomatic patients.[3,6,7]

The aim of this review is to describe the clinical, radiological, pathological findings, progress, and follow-up of 20 children diagnosed with CLM in the University Hospital Parc Taulí between 2006 and 2016.

We reviewed the medical records of 20 children with CLM: 12 boys (60%) and 8 girls (40%). Twelve children (60%) were diagnosed during the prenatal period. Congenital lung malformations were identified as: pulmonary sequestration: 9 cases, (45%), 7 extralobar (77.7%), 2 intralobar (22.22%); CPAM: 5 cases (25%),

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Table 1
Patient Characteristics.

Patient	Type of Malformation	Age at Diagnosis (Months)	Sex	Diagnosis in Prenatal Period	Clinical Characteristics	Age (Months) at Surgery	Affected Lobe
1	ELPS	0	M	CPAM II vs. PS	Asymptomatic	17	RUL
2	ILPS	0	F	PS	Pneumonia	48	LLL
3	ELPS	204	M	No	Hemoptysis	204 Pulmonary embolization	RLL
4	ELPS + CPAM II	0	M	No	NRD	0	LLL
5	ELPS	0	F	CPAM	Asymptomatic	7	RLL
6	ILPS	0	M	PS	Asymptomatic	17	RLL
7	ELPS	0	F	PS	Bronchitis	15	LLL
8	ELPS	0	M	PS	Bronchitis	16	LLL
9	ELPS	0	F	CPAM vs. PS	Asymptomatic	Clinical and radiological monitoring	LLL
10	CPAM II	8	M	No	Bronchitis	12	RUL
11	CPAM I	0	F	CPAM	Asymptomatic	11	RLL
12	CPAM I	0	F	CPAM	Asymptomatic	12	RLL
13	CPAM II	0	F	CPAM II	Asymptomatic	6–24	RLL
14	BC	17	M	No	Bronchitis	22	RUL
15	AB	20	M	No	Pneumonia	–	RUL
16	AB	9	M	No	Pneumonia	–	LLL
17	AB	4	M	No	Pneumonia	–	LLL
18	CLE	0	F	BA-CPAM	Asymptomatic	12	LUL
19	CLE	0	M	BA vs. CLE	Asymptomatic	24	RUL
20	CLE	0	M	No	NRD	1	LUL

BA: bronchial atresia; BC: bronchogenic cyst; CLE: congenital lobar emphysema; CPAM: congenital airway lung malformation; ELPS: extralobar pulmonary sequestration; ELPS: intralobar pulmonary sequestration; LLL: left lower lobe; LLL: left upper lobe; NRD: neonatal respiratory distress; PS: pulmonary sequestration; RLL: right lower lobe; RUL: right upper lobe.

subtype I: 3 (60%), subtype II: 2 (40%), 1 associated with extralobar sequestration; bronchial atresia: 3 (15%); bronchogenic cyst: 1 (5%); congenital lobar emphysema 3 (15%). Clinical presentation in the postnatal period was as follows: 2 patients had neonatal respiratory distress; 9 patients (45%) were asymptomatic; 3 (15%) had recurrent lung infections; and 6 (30%) had episodes of bronchitis. Diagnostic fiberoptic bronchoscopy was performed in the 3 cases of bronchial atresia and lesion sites were distributed as follows: 5 in RUL, 7 in LLL, 6 in RLL, and 2 in LUL. Sixteen were treated with surgery (80%) at an average age of 14 months (2 days-48 months) (Table 1).

In the group of 9 asymptomatic patients (45%), 8 were treated with surgery, and of these 2 had congenital lobar emphysema, 3 CPAM, and 3 pulmonary sequestration. One patient with CPAM type II required 2 surgical interventions (6 and 24 months of age), with subsequent clinical and radiological follow-up, in view of persistent CPAM. The asymptomatic patient who was not treated with surgery was a case of extralobar pulmonary sequestration that was managed conservatively with a favorable outcome.

The most common lung malformations in our series were CPAM and pulmonary sequestration. Prenatal ultrasonography provided the diagnosis in more than half of the cases. The chest X-ray of most patients diagnosed by prenatal ultrasonography was normal in the neonatal period, and diagnosis was confirmed in all patients with chest CT. Eighty percent of the children were treated with surgery, with no postoperative complications. The remaining patients continued to progress favorably in the 10 years of follow-up.

As reported in other reviews, the diagnosis of this disease has increased with the use of prenatal ultrasound, while chest CT remains the gold standard diagnostic method. According to the literature, the management of asymptomatic patients is still controversial, and many authors are inclined toward early elective surgery to avoid the risk of complications including infections and the risk of malignant transformation.[8,9] In our review, about half the patients were asymptomatic, and most were treated with surgical resection.

Criteria for surgery have been identified in patients at higher risk, while conservative management may be selected for lower-risk patients.[3,10] Proponents of non-surgical treatment report that some CLM can spontaneously resolve in the postnatal period, but the prevalence of complications is unknown.[6]

The current trend is toward performing minimally invasive pulmonary resections via video-assisted thoracoscopy, due to the low morbidity and good postoperative results offered by this procedure.[11,12]

The involvement of a multidisciplinary team is of vital importance in the management of these patients, and the most appropriate treatment should be selected according to the type of malformation and the presence of symptoms, taking into consideration the risk of complications in each case and the benefit of surgery.

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The Effect of a Basic Basket on Tuberculosis Treatment Outcome in the Huambo Province, Angola

Efecto de una cesta básica alimentaria en el resultado del tratamiento de la tuberculosis en la provincia de Huambo, Angola

Dear Director:

Tuberculosis (TB) continues to be a worldwide health problem and is the greatest infectious cause of death among young people and adults.[1]

Although TB treatment is free, patients will often stop treatment due to the inability of the patient and his/her relatives to provide the resources necessary to survive.[2] Studies have shown that poverty and TB are closely linked, since the conditions under which poor populations live make them prone to the onset of the disease.[1,3] The economic impact of TB arises from the fact that the majority of those affected are part of the labour force.[1] Therefore, incentives have been employed by national TB control programs as a means of encouraging patients to adhere to treatment, helping to minimize the costs associated with the disease, as well as providing support to their families during the course of treatment.[4]

The consequences of the armed conflict that has occurred in Angola for about three decades has affected mainly the migration of the rural population to other regions, and the destruction of social infrastructures and traditional systems of economic activity.[5] In addition, the destruction of roads has led to the shortage of basic and other essential goods, giving rise to dramatic humanitarian situations.[6] In this sense, the government has focused its attention on the affected populations, with the support of international institutions, mainly the United Nations Organizations (UN), through the World Food Program (WFP).[6] Throughout the 1990s, WFP provided Angola with logistical support for intervention among populations in critical areas.

Since 2002, after the signing of the peace agreement in Angola,[7] WFP has developed a humanitarian aid plan for TB patients hospitalized at the Huambo Sanatorium hospital and those who were being treated at the Anti-Tuberculosis Dispensary (ATD) outpatient clinic. This aid lasted 6 years and consisted in the distribution of a basic food basket that patients took home once a month during the outpatient treatment period. Inpatients received the prepared food at the hospital.

The program's implementation had as its purpose increasing the adherence of TB patients to the Directly Observed Treatment (DOT) and improving the success rates of anti-tuberculosis treatment.[8]

Due to lack of funding, WFP was forced to end humanitarian aid in Angola by the end of 2006, ahead of schedule. The government then assumed responsibility for providing the basic food basket to patients through the Ministry of Planning and the Ministry of Social Assistance and Reintegration (MINARS) until 2008.[9] Since 2009 there has been no further incentive for TB patients.

The objective of this study was to evaluate the effect of incentives (basic food basket) on the treatment outcome of patients with TB in the ATD, Huambo.

A retrospective study is carried out. The study included 19 739 cases of TB that were treated in the period between 2002 and 2014, in the ATD, Huambo. The study time was divided into two periods, the first one referring to the years 2002–2008, the period in which the basic baskets were distributed, and the second period referring to the years 2009–2014, during which there were no basic baskets. Data on the total TB cases and treatment outcomes were collected through the annual ATD reports, Huambo.

Treatment results were defined according to the WHO guidelines.[10]

The data available was analyzed through descriptive statistics. Mann–Whitney *U* test was used for group comparison.

Table 1
Results of the Patients' Treatment, Diagnosed With TB in the DAT, Huambo, During the Period From 2002 to 2014.

Year	Number of cases	Success (%)	Failure (%)	Deceased (%)	Abandonment (%)	Transferred (%)
With basic basket						
2002	1591	1421 (89.3)	4 (0.3)	53 (3.3)	110 (6.9)	3 (0.2)
2003	1694	1551 (91.6)	5 (0.3)	32 (1.9)	98 (5.8)	8 (0.5)
2004	1563	1395 (89.3)	7 (0.4)	41 (2.6)	111 (7.1)	9 (0.6)
2005	1664	1465 (88.0)	9 (0.5)	59 (3.5)	118 (7.1)	13 (0.8)
2006	1143	988 (86.4)	4 (0.3)	40 (3.5)	105 (9.2)	6 (0.5)
2007	1590	1328 (83.5)	3 (0.2)	50 (3.1)	200 (12.6)	5 (0.3)
2008	1567	1369 (87.4)	6 (0.4)	37 (2.4)	144 (9.2)	11 (0.7)
Without basic basket						
2009	1122	569 (50.7)	15 (1.3)	51 (4.5)	462 (41.2)	25 (2.2)
2010	1780	1042 (58.5)	24 (1.3)	76 (4.3)	617 (34.7)	21 (1.2)
2011	1871	1325 (70.8)	18 (1.0)	67 (3.6)	447 (23.9)	19 (1.0)
2012	1505	914 (60.7)	10 (0.7)	57 (3.8)	508 (33.8)	16 (1.1)
2013	1425	859 (60.3)	8 (0.6)	38 (2.7)	502 (35.2)	18 (1.3)
2014	1224	760 (62.1)	9 (0.7)	34 (2.8)	401 (32.8)	20 (1.6)